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Pulmonary Echinococcosis: A Pediatric Disease of the Southwestern United States

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ABSTRACT. Three cases of pulmonary echinococcosis in native American children emphasize the need to consider the diagnosis in the differential diagnosis of mass lesions of the chest when the appropriate environmental conditions are present. Two children presented with a non-productive cough, while one was asymptomatic. Surgery is the treatment of choice for pulmonary echinococcal cysts. All three children did well after thoracotomy, and one child has a normal roentgenogram six years after surgery. Recent evidence seems to indicate that transmission of the *Echinococcus* among native Americans in the Southwest is increasing, and thus human echinococcal disease may be of increasing clinical importance. *Pediatrics* 65:1003-1006, 1980; *Echinococcus granulosus*, *pulmonary hydatid cysts*, *pulmonary masses*, *native Americans*.

Hydatid disease (echinococcosis), a parasitic infestation of animals transmissible to man, is a major health problem in many parts of the world where livestock, especially sheep, are kept.¹ The disease is considered to be rare in the United States, and most reported cases have occurred in immigrants.² Locally acquired echinococcosis has now been documented in American Indians living in Arizona and New Mexico.³ The two-state region is the most recently disclosed extension of the parasite's range, and it appears that transmission to humans may be increasing.⁴ In other parts of the world, 50% to 80% of patients with pulmonary echinococcal disease have been children.⁵ We are unaware of any previous reports of American children with pulmonary echinococcosis.

We present three native American children with pulmonary echinococcosis treated at our institution. We report these cases to emphasize the need

to consider this diagnosis in children with mass lesions of the chest.

CASE REPORTS

Case 1

A 5-year-old Navajo boy from Chinle, AZ, was referred to Bernalillo County Medical Center because of an abnormal chest roentgenogram which was obtained during a routine school physical examination (Fig 1). The patient was asymptomatic and had been in good health all his life. He frequently visited his grandmother who was a sheepherder. There were many stray dogs in the area.

No abnormalities were noted on physical examination. Laboratory data showed a normal complete blood count with the exception of a sedimentation rate of 51 mm/hr. Tomograms of the chest revealed a solid round lesion in the superior segment of the right lower lobe. A liver-spleen scan was interpreted as normal. At thoracotomy, an intact 5x5-cm white cyst was excised by wedge resection from the right lobe. Touch prep of the fluid showed the typical proglottids of the echinococcus (Fig 2). The patient made an uneventful recovery.

Case 2

A 2½-year-old girl from Santo Domingo, NM, was admitted with a two-day history of fever, irritability, and nonproductive cough. She had been entirely well until this episode. Her family had a small farm on which sheep and other animals were raised. There were many stray dogs in the area. Physical admission examination revealed a well-developed, well-nourished girl in mild respiratory distress. Vital signs were unremarkable except for a temperature of 39 C. Upon examination of the chest, scattered rales were heard at the left base. The remainder of the physical examination was normal. Laboratory data were within normal limits with the exception of a sedimentation rate of 38 mm/hr. Chest roentgenogram revealed an oval cystic lesion in the right middle lobe and a left lower lobe infiltrate (Fig 3). Ampicillin was administered. Skin tests for tuberculosis, coccidiomycosis, and histoplasmosis were negative. Serum echinococcal hemagglutination-inhibition titers were positive at 1:4,096. Liver-spleen scan and brain scan were interpreted as normal. At thoracot-

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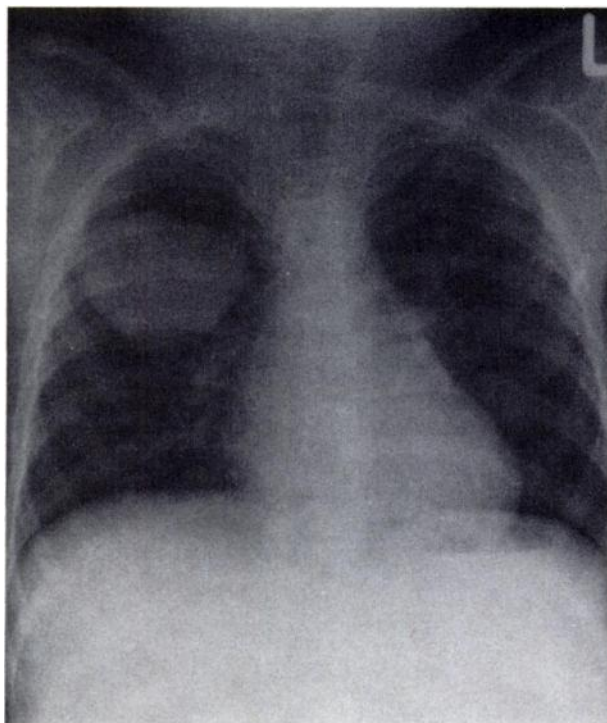


Fig 1. Roentgenogram of case 1 showing mass in right lower lobe.

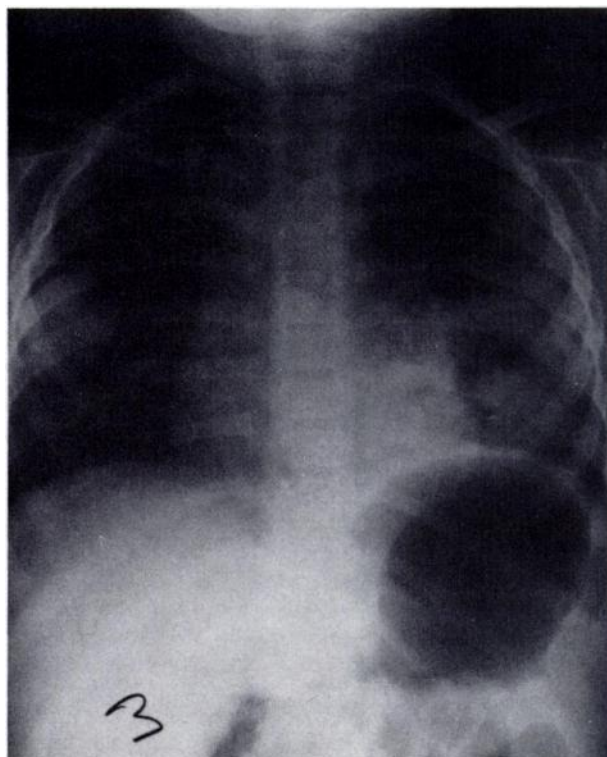


Fig 3. Roentgenogram of case 2 showing mass in right middle lobe and left lower lobe infiltrate.

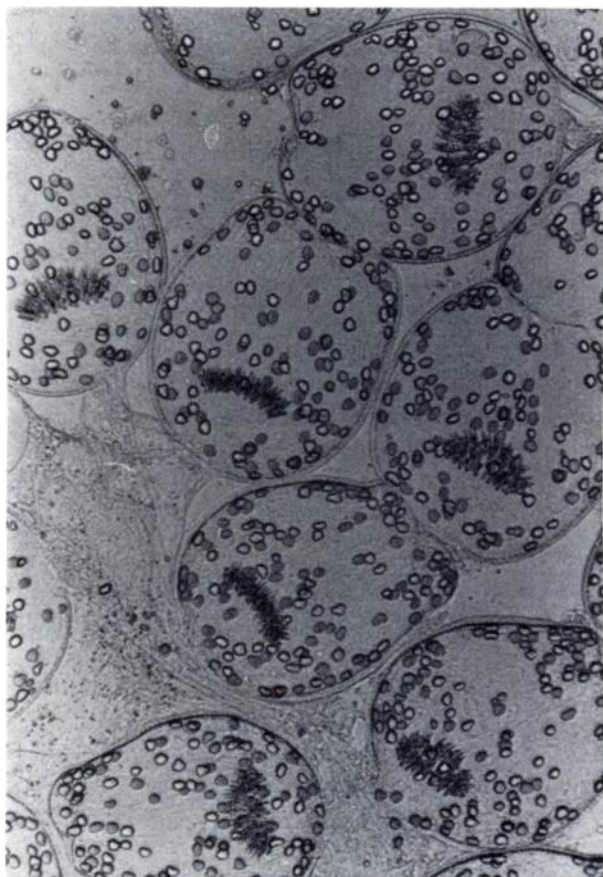


Fig 2. Touch prep of fluid from cyst showing typical proglottids of the echinococcus.

omy, a 3x4-cm cyst was removed intact from the right middle lobe. The fluid showed the typical proglottids of the echinococcus. Subsequently, she developed several cysts of the liver which were removed. Six years later her chest roentgenogram remains normal.

Case 3

A 5-year-old Navajo girl was admitted because of a growing mass lesion of the right upper lobe noted five months previously. She had a low grade fever and persistent nonproductive cough of three months duration. There were three family members with tuberculosis. Despite the fact that sputum and gastric specimens were negative for acid fast bacilli, the patient was placed on isoniazid (INH) and ethambutol. The mother admitted that the family occasionally ate poorly cooked lamb's meat. Physical examination was unremarkable, as was a complete blood count. Serum echinococcal hemagglutination-inhibition titer was positive at 1:256. At operation, an intact cyst was removed from the right upper lobe. The laminated acellular basophilic layer of the cyst was consistent with echinococcal disease. Despite multiple stains, no organisms could be visualized. The patient made an uneventful recovery but has been lost to follow-up.

DISCUSSION

The minute adult tapeworm, *Echinococcus granulosus*, lives in the intestines of canine animals, especially dogs. When the eggs are ingested by

people, they hatch in the duodenum, liberating larvae that penetrate the intestinal mucosa and reach the liver, lung, and other tissues by hematogenous and lymphatic routes.⁶ Larvae that survive usually develop into unilocular cysts. As the hydatid cyst grows in size, the lung reacts by forming a connective tissue layer, the pericyst which surrounds the parasite. It is estimated that the cyst grows 1 to 2 cm a year.⁷ In a recent survey of 114 children treated for hydatid disease from Australia, 62 had cysts in the lungs, 40 had cysts in the liver, and 12 had cysts in both lungs and liver.⁸ Other organs—brain, kidney, and spleen—are rarely involved. In children with pulmonary cysts, the presenting symptoms may be hemoptysis, cough, or acute chest pain. Approximately 5% of patients may be asymptomatic as our first patient was.⁹ Rupture of a pulmonary cyst may be associated with an abrupt onset of cough, fever, and occasionally expectoration of scoleces. An acute anaphylactic reaction may ensue from spillage of the cyst contents.¹⁰

Hydatid cysts of the lung are usually solitary (75%) and usually in the right lung.¹⁰ The cysts are oval or spherical in shape. Calcification of the cyst wall is rarely present on roentgenogram.⁷ A floating membrane called the water-lily sign is seen with an air-fluid level in ruptured cysts. Pneumothorax, hydropneumothorax, and empyema may be seen when the cyst ruptures into the pleural space.

The differential diagnosis considered in these cases of isolated pulmonary cyst included bronchiogenic cyst, hamartoma, postinflammatory nodule, granuloma, and echinococcal cyst. In all three cases, because of the epidemiologic circumstances, echinococcal disease was considered the most likely in possibility.

Roentgenographic signs are generally diagnostic in countries where hydatid disease is common. However, in the United States where the disease is uncommon, a high index of suspicion is required when the appropriate environmental conditions are present. In regard to laboratory diagnosis, the Cassoni skin test is no longer performed due to the high percentage of false-positives. Eosinophilia is a non-specific finding of many parasitic infections, and thus not pathognomonic of echinococcal infection. Additionally, the absence of eosinophilia does not definitely rule out echinococcosis. The indirect hemagglutination test appears to be the most reliable diagnostic technique and is positive in about 85% of the cases.¹¹ This titer can be obtained at the Center for Disease Control in Atlanta.

Operation is the treatment of choice for pulmonary hydatid cysts. A three-year trial of high dose mebendazole therapy for advanced alveolar hydatid disease in four patients was recently reported.¹² The

results were encouraging but not conclusive, and the exact role of drug therapy is yet to be defined. Transthoracic needle aspiration of a suspected cyst must never be attempted, as spillage of its contents could cause severe anaphylactic reaction. At thoracotomy, the cyst is located by gentle palpation of the lung. A wedge resection is performed, using the stapling instrument, and the cyst is excised, including a margin of about 1 cm of normal lung tissue. In the alternative technique, clamps, instead of staples, are applied to the lung, the cyst is excised, and the lung margin is sealed by a running suture. Some surgeons prefer to inject the cyst with hypertonic saline before excision. In 80 children from Australia operated upon for pulmonary cysts, there were no deaths; however, there were six children with recurrences who required a second operation. The only deaths in the above series occurred with cysts of the brain.⁸

Existing evidence suggests that *E granulosus* was first introduced into the United States from Europe prior to 1900 when it was reported frequently in swine populations in the Southeastern states.¹³ More recently, spread has occurred to several Western states where the parasite now has been found in sheep. Human populations at unusual risk in the Western United States are sheep ranchers, including Basque-Americans in California, Mormons in Utah, and now the Navajo, Zuni, and Santo Domingo Indians in Arizona and New Mexico, whose husbandry practices permit dogs to eat the uncooked parts of sheep.

Recent epidemiologic surveys indicate that the dogs responsible for human infection may have become infected from both sheep raised locally in rural areas of the Navajo reservation and from off reservation sheep acquired at local trading posts.¹⁴ It is clear that *Echinococcus* is endemic at low prevalence in the sheep dog cycle in rural areas of the Indian reservations and that if present trends continue it will be of growing clinical importance. Pulmonary echinococcosis should be considered in the differential diagnosis of mass lesions of the chest, especially in native American patients from the Southwestern United States.

ACKNOWLEDGMENT

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In the article "*Haemophilus influenzae* Type b in a Day Care Center: Relationship of Nasopharyngeal Carriage to Development of Anticapsular Antibody" by Granoff et al (*Pediatrics* 65:65-68, 1980), on p 66 in Table 2 the geometric mean antibody level for adult blood bank donors should read 698 not 459.

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In the article "Morning Plasma Cortisol Levels in Infants Treated with Topical Fluorinated Glucocorticosteroids" by Weston et al (*Pediatrics* 65:103, 1980) an error has been noted in the description of the method used for cortisol assay. Samples were assayed by the competitive-protein binding radioassay of Murphy, which permits measurement of cortisol in 0.1-ml aliquots of plasma (0.2 ml for a duplicate determination) with a coefficient of variation of 7%.^{1,2} The Spencer-Peet fluorometric method, noted incorrectly in the text, would require a much larger volume of plasma than that obtained from the infants. References: (1) Murphy BEP: Some studies of the protein-binding of steroids and their application to the routine micro and ultramicro measurement of various steroids in body fluids by competitive protein binding radioassay. *J Clin Endocrinol Metab* 27:973, 1967; (2) Morris HG, DeRoche G, Winkler SM, et al: Effect of oral prednisone on the measurement of plasma steroid concentrations by the competitive protein binding radioassay. *J Pediatr* 85:248, 1974.

In the article "Human Milk Banking" (*Pediatrics* 65:854, 1980) the name of the principal author of the Committee on Nutrition, Claude C. Roy, MD, was inadvertently omitted from the list of committee members.

In the abstract "Differentiating Minimal Brain Dysfunction and Temperament" (*Pediatrics* 65:A40, 1980) the authors' names were given incorrectly. The sentence should read abstracted from W. B. Carey, S. C. McDevitt, and D. Baker (*Dev Med Child Neurol* 21:765, 1979).

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